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CASE REPORT

Bladder Outlet Obstruction due to an Unusual Prostatic Midline Cyst in an Adolescent Male

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Most prostatic midline cysts, i.e., Müllerian duct cysts or prostatic utricular cysts, probably result from incomplete regression of the Müllerian duct remnant. Congenital cysts may be associated with other urogenital anomalies. Related symptoms include bladder outlet obstruction, hemospermia, ejaculatory-fertility impairment, and recurrent epididymitis. We report an adolescent male who presented with difficulty urinating that lasted for 2 months. After a complete survey with a urodynamic study, ultrasonography, urethrography, urethrocystoscopy, and computed tomography, a prostatic midline cyst with bladder outlet obstruction was identified. Using transurethral cyst unroofing, urination conditions improved according to the International Prostate Symptom Score and a uroflow study. Although there are various surgical interventions for a prostatic midline cyst near the urethra, endoscopic procedures are a safe and elegant alternative compared with other procedures.

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1. Introduction

Most prostatic midline cysts are an uncommon isolated congenital anomaly of males and are derived from the caudal ends of fused Müllerian ducts.[1] They can be classified into two interchangeable designations: Müllerian duct cysts and prostatic utricle cysts. Prostatic midline cysts occur in male subjects of all ages but are especially common among males in their 3rd and 4th decades.[2] Congenital cysts are usually associated with other congenital abnormalities of the urinary tract. On the other hand, acquired cysts can also be attributable to the posterior urethritis and verumontanitis. The clinical signs and symptoms depend on the size of the cyst and the presence of an infection. We present a case of an adolescent with severe difficulty urinating and bladder outlet obstruction due to a prostatic midline cyst, and we review the literature.

2. Case Report

A 19-year-old male adolescent presented with a 2-month history of dysuria. He had previously been well but had been having unprotected sexual intercourse for approximately 3 months. The condition of dysuria slightly improved after ejaculation. The patient denied any other lower-urinary-tract symptoms, a history of trauma, or recent medication history. A digital rectal examination revealed a normal-sized prostate with no hard nodules

or tenderness. Normal erectile function and ejaculation were described by the patient. Semen characteristics and sperm motility were not affected. A complete urinalysis showed no pyuria or hematuria, and there was no growth of aerobic bacteria in culture. The prostate-specific antigen level was 1.2 ng/mL. The International Prostate Symptom Score was 22 with predominant obstructive scores. A urodynamic study showed a high voiding pressure (>50 cmH_2O) and a low flow rate (Qmax of <10 mL/s), and was compatible with normal detrusor function with bladder outlet obstruction. A cystic-like lesion in the midline of the prostate ($2\times1.8\times4$ cm) was observed on ultrasonography (Figure 1). Urethrography was performed by a clinical surgeon. Contrast medium was transfused retrograde into the urinary bladder, and stretching of the prostatic urethra by the prostatic cyst was identified (Figure 2). Surveillance by computed tomographic imaging 2 months after presentation of difficult urination revealed a low-density cystic component lesion in the central zone of the prostate with focal peripheral enhancement. Engorgement of the bilateral seminal vesicles was also observed (Figure 3).

Because clinical image findings were consistent with a prostatic midline cyst with bladder outlet obstruction, the patient was admitted for surgical intervention. We preferred transurethral cyst unroofing because the cyst was near the urethra. During the procedure, cystourethroscopy revealed that there was a cystic lesion at the verumontanum with bladder outlet obstruction. Some brown fluid was exuded from the cyst after the roof was broken with a cutting loop. We also identified the openings of the ejaculatory duct and protected them. The inside of the cyst had a smooth surface and mucosa-like urothelium (Figure 4). The pathological diagnosis of a cystic roof was based on fibrotic tissue lined by the urothelium with chronic inflammatory cell infiltration. The patient's postoperative condition was good. The Foley catheter was removed 24 hours after surgery. Two weeks later, repeat ultrasonography revealed no cystic lesion in the prostate. The condition of the bladder outlet obstruction improved according to the International Prostate Symptom Score and a uroflow study (Qmax of 16 mL/s). Normal

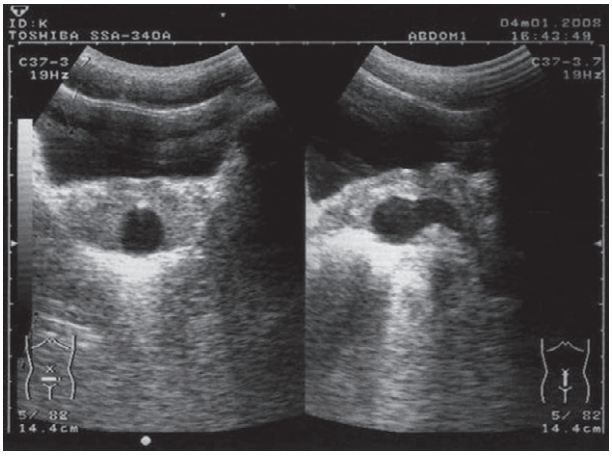

Figure 1 Ultrasonogram shows a cystic-like lesion on the midline of the prostate.

Figure 2 Retrograde urethrogram shows stretching of the prostatic urethra by a prostatic cyst. There was no contrast filling in the cyst during contrast medium retrograde transfusion.

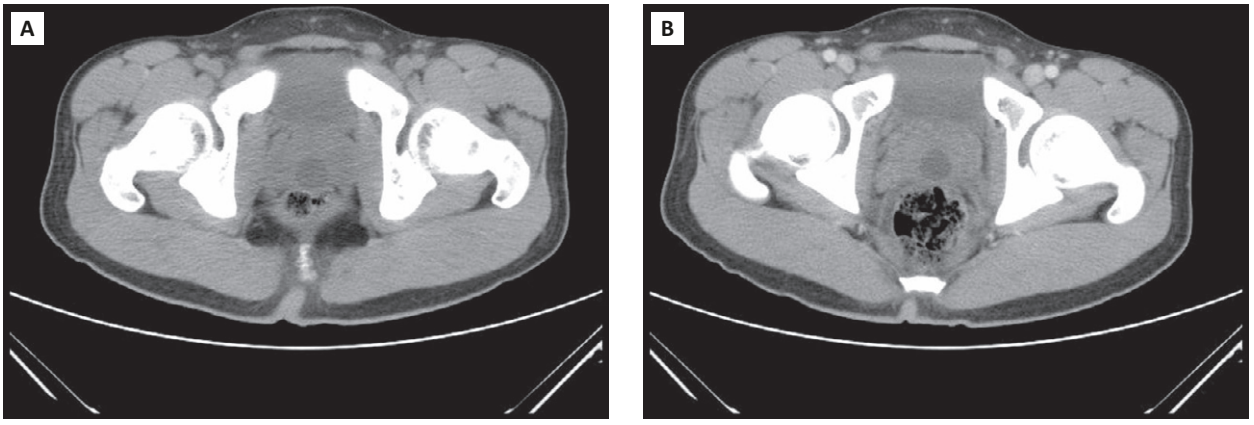

Figure 3 Abdominal computed tomogram showing a low-density cystic component lesion in the central zone of the prostate with focal peripheral enhancement: (A) before enhancement and (B) after enhancement.

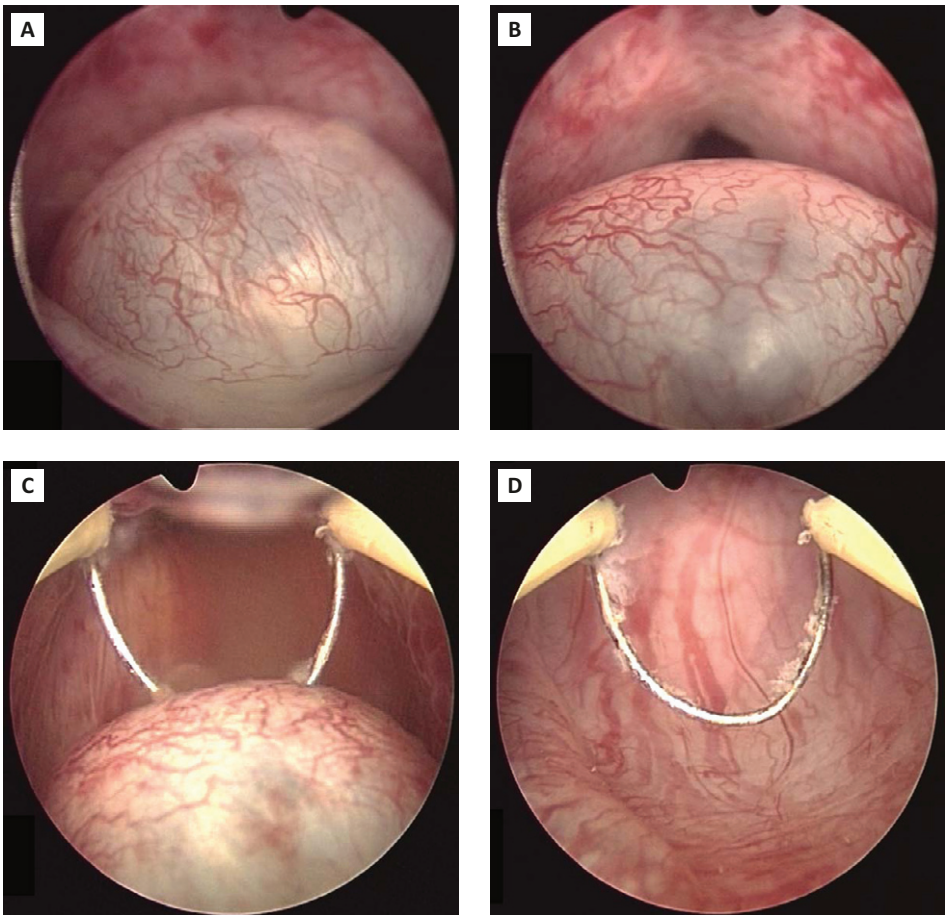

Figure 4 (A) A cystic lesion at the verumontanum. (B) Bladder outlet obstruction due to the cyst. (C) Brown exudate from the cyst after the roof was broken with a cutting loop. (D) The inside of the cyst showing a smooth surface and urothelium-like mucosa.

post-voiding residual urine (35 mL) was measured by ultrasonography. In a 6-month follow-up after the operation, the patient appeared well with normal voiding and sexual function.

3. Discussion

It was originally believed that prostatic midline cysts were probably caused by incomplete regression of the Müllerian duct remnant. Anatomically, the cyst is derived from the caudal end of the fused Müllerian duct, originating at the level of the verumontanum and extending medially and posteriorly above the prostate gland.[1] The terms "Müllerian duct cyst" and "prostatic utricle cyst" are used interchangeably for a midline cyst of the prostate, leading to some confusion. Based on previous embryological examinations, utricular cysts are of endodermal origin, they contain no spermatozoa, and they are located near the verumontanum, whereas Müllerian cysts are of mesodermal origin, they contain spermatozoa, and they are located more posterior and nearer to the prostate base. Therefore, these two cysts are differentiated according to whether or not there is communication between the cyst and urethra. A diagnosis of Müllerian duct cyst appears to be made mainly by imaging evidence or surgical findings that confirm no relationship between the cyst and prostatic urethra. However, a recent study of histological and immunohistochemical examinations of the epithelium lining prostatic midline cysts indicated that its characteristics are identical to those of the prostatic utricle. Since there is no evidence that these cystic lesions originate from the Müllerian duct remnant, it has been suggested that they be re-termed "cystic dilation of the prostatic utricle" or "utricular cyst", depending on whether a communication to the prostatic urethra is present or absent.[3] Although prostatic midline cysts are an uncommon isolated congenital anomaly in males and are usually associated with other congenital urogenital abnormalities, such as hypospadias, undescended testes, and unilateral renal agenesis,[4] they can also be acquired due to obstruction of the utricular orifice. Trauma and low-urinary-tract infection, such as posterior urethritis and verumontanitis, are the most common causes.

Other differential diagnoses of cystic prostatic lesions near the midline location include an ejaculatory duct cyst, vas deferens cyst, transurethral resection of the prostate defect, and prostatic abscess.[5] Several approaches have been used to make a diagnosis, including a digital rectal

examination, cystoscopy, retrograde urethrography, and seminal vesiculography. More recently, transrectal ultrasound, computed tomographic, and magnetic resonance imaging have been used in clinical assessments and they provide a detailed demonstration of the zonal anatomy and a large field of view in all three orthogonal planes.[6] Although the range of noninvasive radiologic techniques is extensive, invasive procedures, such as needle aspiration of the cyst with direct injection of contrast medium or surgical exploration, remain essential for further distinction. If there is no sperm, a prostatic midline cyst is favored.

The clinical signs and symptoms are dependent on the size of the cyst and the presence of infection and abnormalities. Although almost 60% of adults diagnosed with a prostatic midline cyst do not experience any cyst-related symptoms,[7] some clinical presentations, including dysuria, lower-urinary-tract symptoms, bladder outlet obstruction, suprapubic or rectal pain, hematuria, hemospermia, ejaculatory-fertility impairment, and recurrent epididymitis, may trouble some patients. Cornel et al. demonstrated that the ejaculatory duct runs in an almost straight course from the prostatic base to the verumontanum and, therefore, it is believed that a midline prostatic cyst could be one of the causes of ejaculatory duct obstruction by lateral compression of the ejaculatory duct.[8]

Surgical therapy is reserved for symptomatic cysts. Previously, various open surgical approaches were performed including suprapubic excision, transperineal or transrectal drainage, posterior sagittal transanal approaches, and various transtrigonal techniques, all of which are accompanied by limited anatomical visualization and potentially significant morbidity from iatrogenic damage of adjacent structures.[9] However, minimally invasive procedures have recently been described, such as ethanol sclerotherapy (transperineal or transrectal) and endoscopic techniques. Transurethral resection or unroofing of a prostatic midline cyst near the urethra offers a safe and elegant alternative to all other methods by improving visualization of the structures and potentially reducing morbidity from iatrogenic injury.[10] However, some late complications, including urethral stricture and external sphincter injury should be closely followed-up after the transurethral procedure. In our study, after ruling out a neurogenic bladder by a urodynamic study, the male adolescent was diagnosed with bladder outlet obstruction due to a rare prostatic midline cyst. We believe that it should be termed a "utricular cyst" because there was no communication between the cyst and prostatic urethra. The patient has been doing well with normal voiding and sexual function after transurethral cyst unroofing.

In conclusions, prostatic midline cysts are rare urogenital anomalies in males and are associated with bladder outlet obstruction at a young age. Endoscopic techniques are recommended as the first-line treatment for symptomatic patients with a cyst near the urethra.

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